Case Report

Bilateral High-Grade Sertoli–Leydig Cell Tumors with Gallbladder Metastasis Presenting as Obstructive Jaundice

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Abstract

Objective: To report a rare case of bilateral high-grade Sertoli–Leydig Cell Tumor (SLCT) with gallbladder metastasis presenting as obstructive jaundice.

Methods: This is a case report of a 52-year-old woman presenting with gastrointestinal and hepatobiliary symptoms. Clinical evaluation included laboratory testing, imaging studies (ultrasound, Multislice Computed Tomography [MSCT], and Magnetic Resonance Cholangiopancreatography [MRCP]), surgical intervention, histopathological examination, and immunohistochemical analysis.

Case: The patient presented with heartburn, jaundice, nausea, vomiting, pale stools, and dark urine. Laboratory findings revealed elevated liver enzymes, mildly increased carcinoembryonic antigen (CEA), and normal cancer antigen 125 (CA-125). Imaging studies identified bilateral ovarian masses, while MRCP demonstrated multiple gallbladder stones with features of cholangitis and obstructive jaundice. The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy and cholecystectomy. Histopathological evaluation revealed poorly differentiated Sertoli–Leydig cell tumors in both ovaries, characterized by diffuse and microtubular growth patterns, nuclear pleomorphism, and mucin-containing cytoplasm with occasional signet-ring cell morphology. Immunohistochemistry showed positivity for calretinin and inhibin, and negativity for epithelial membrane antigen (EMA) and cytokeratin (CK), confirming the diagnosis of high-grade SLCT. Examination of the gallbladder demonstrated metastatic deposits consistent with an ovarian primary tumor.

Conclusion: Sertoli–Leydig cell tumors are rare ovarian neoplasms, accounting for less than 0.5% of all ovarian tumors. Prognosis is largely determined by tumor grade and histological subtype, with high-grade variants associated with more aggressive behavior and poorer outcomes. This case highlights an exceptionally rare presentation of bilateral high-grade SLCT with gallbladder metastasis leading to obstructive jaundice. Recognition of such atypical metastatic patterns is important for accurate diagnosis and management.

Keywords: metastasis vesica fellea, obstructive jaundice, Sertoli-Leydig Cell Tumor.

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INTRODUCTION

Sertoli–Leydig cell tumors (SLCTs) of the ovary are classified as sex cord–stromal tumors (SCSTs), which are composed of sex cord elements (granulosa and Sertoli cells) and stromal components (fibroblasts, theca cells, and Leydig cells).[1,2] Sertoli and Leydig cells are normally found in the testes,[3] where they are responsible for androgen production; therefore, SLCTs are also referred to as androblastomas.[4] Some SLCTs, however, may exhibit hyperestrogenism.[5] SCSTs account for approximately 3–7% of all ovarian tumors, while SLCTs are rare, with an annual incidence of 2.1 per 100,000 women and representing only 0.2% of all ovarian tumors.[1,2,6] SLCTs can occur across a wide age range, from 4 to 82 years, with 26.6%–56% of cases reported in postmenopausal women.[7-10] Histologically, SLCTs are classified into well-differentiated (grade 1), moderately differentiated (grade 2), and poorly differentiated tumors (grade 3), which may include heterologous, retiform, or mixed components.[11]

SLCTs are typically diagnosed at an early stage, are usually unilateral, and often have a favorable prognosis.[4,12,13] In contrast, advanced-stage or high-grade (grade 3) tumors are associated with poorer outcomes, including higher recurrence rates and significantly reduced overall survival. High-grade SLCTs have been reported to have a hazard ratio of 14.25 (95% CI: 1.88–108.0; log-rank p = 0.010) compared with well- or moderately differentiated tumors.[14] Metastases have been reported in the lungs, scalp, supraclavicular lymph nodes, and liver.[15] Bilateral involvement, including metachronous presentation, has also been described.[16]

This case report describes a rare presentation of bilateral high-grade Sertoli–Leydig cell tumor involving both ovaries, characterized by heterologous mucinous differentiation with occasional signet-ring cell morphology, and metastasis to the gallbladder wall.

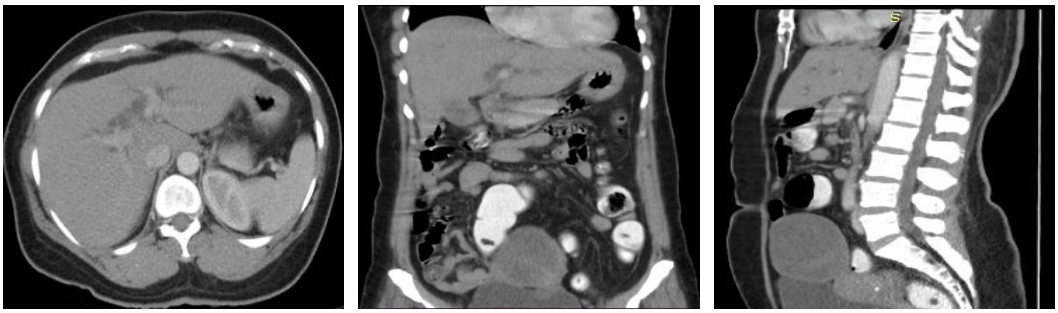
METHODS

This is a case report of a 52-year-old woman presenting with obstructive jaundice and bilateral ovarian masses. Clinical evaluation included history taking, physical examination, laboratory tests (liver function and tumor markers: CEA and CA-125), and imaging studies, including ultrasound, Multislice Computed Tomography (MSCT), and magnetic resonance cholangiopancreatography (MRCP). The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy and cholecystectomy. Histopathological examination and immunohistochemical analysis (calretinin, inhibin, epithelial membrane antigen [EMA], and cytokeratin [CK]) were performed to establish the diagnosis. Written informed consent was obtained, and patient confidentiality was maintained.

CASE

A 52-year-old woman presented with heartburn one week prior to hospital admission. Associated symptoms included jaundice (icteric sclera), nausea, vomiting, pale watery stools for five days, and tea-colored urine. Physical examination of the abdomen revealed epigastric tenderness without a palpable mass. On admission, liver function tests showed total bilirubin of 10.21 mg/dL, direct bilirubin 7.35 mg/dL, and indirect bilirubin 2.66 mg/dL. Transaminase levels were elevated (AST/SGOT 119 U/L, ALT/SGPT 252 U/L). Serum carcinoembryonic antigen (CEA) was 4.71 µg/L, while cancer antigen 125 (CA-125) was within normal limits at 6.40 U/mL.

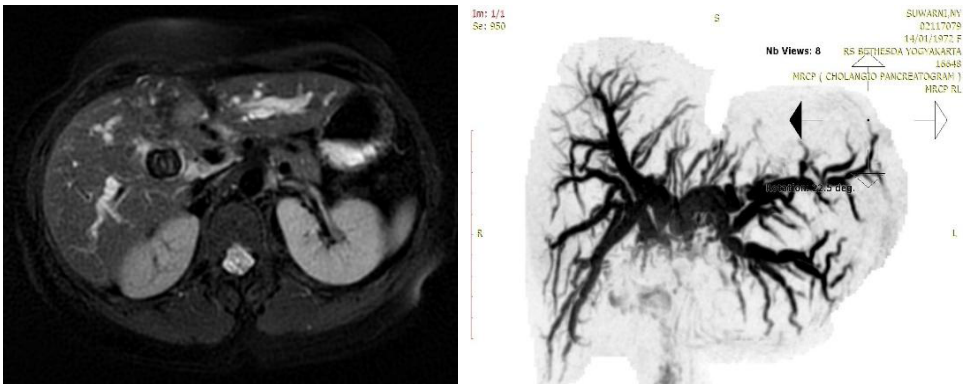
Abdominal ultrasound suggested a cystic-solid parametrial mass suspected to originate from the ovaries, with features suspicious for malignancy (O-RADS 4). Evaluation of the right hypochondriac region demonstrated cholelithiasis and cholecystitis. Multislice computed tomography (MSCT) of the abdomen revealed two main findings (Figure 1). First, a mass measuring approximately 5–6 cm was identified in the anterosuperior and posterior uterus, displacing the sigmoid colon, consistent with an ovarian mass without evidence of regional organ invasion. Second, cholelithiasis was associated with dilation of the intrahepatic biliary system.



A B C

Figure 1. MSCT A. Axial sequence pre-contrast intrahepatic bile duct dilatation (blue arrow). B. Coronal sequence 5mm after contrast. Tumor of ovaries, without any adhesion to other organ (green arrow). Bile duct, dilated, walls thickened with radiopaque stone (blue arrow). C. Sagittal sequence 5mm after contrast. Ovaries tumor, shadowy septal image.

Magnetic resonance cholangiopancreatography (MRCP) demonstrated multiple common bile duct stones accompanied by cholangitis and obstructive jaundice (Figure 2). The primary clinical diagnosis was suspected ovarian malignancy, with additional diagnoses of cholelithiasis, cholecystitis, and obstructive jaundice.



A B

Figure 2. MRCP A. Axial sequence. Intrahepatic bile duct dilatation B. Multiple bile duct calculi with intact connective tissue capsule.

The patient underwent total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, and appendectomy. Gross examination revealed the right ovary measuring 8 × 5 × 4 cm and the left ovary 8.5 × 7 × 6 cm. Histopathological evaluation demonstrated a biphasic ovarian tumor. The hypercellular component consisted of poorly differentiated Sertoli–Leydig cells arranged in diffuse and microtubular patterns, with occasional tubule formation. Tumor cells were pleomorphic with marked nuclear atypia and frequent mitotic figures. Some tumor cells contained mucin with eccentrically displaced nuclei, exhibiting a signet-ring appearance. The hypocellular component consisted of loose fibrous stroma containing eosinophilic and mucinous material, with scattered undifferentiated Sertoli–Leydig cells and mucin-containing signet-ring-like tumor cells.[14]

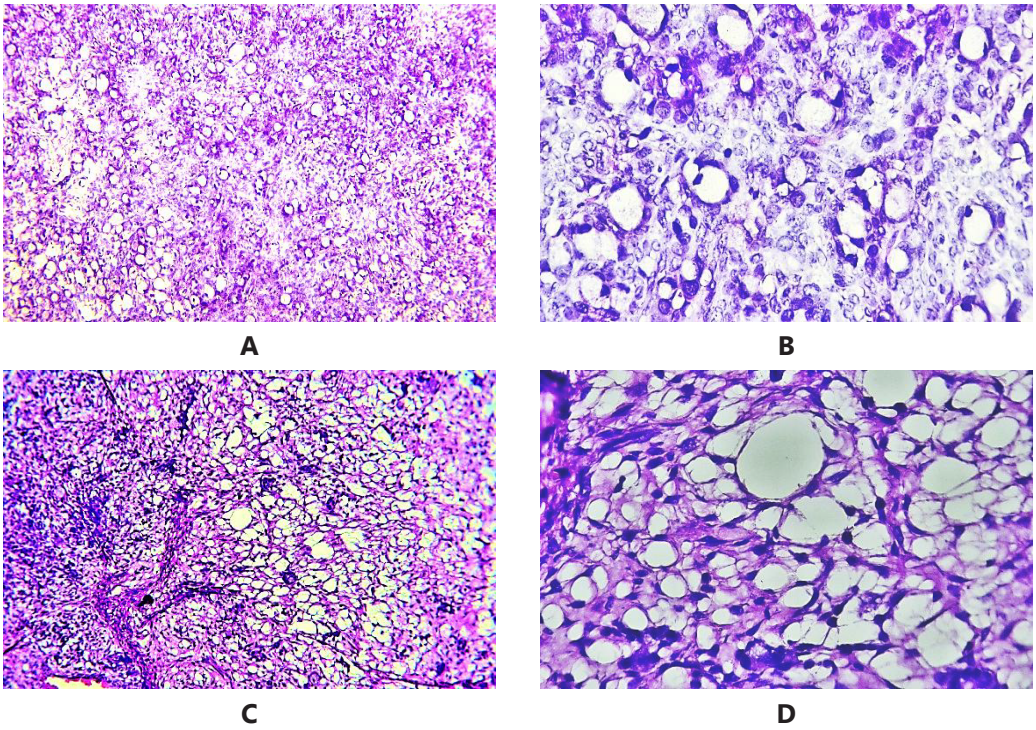


Figure 3. Sertoli- Leydig Cell Tumor. (A, 100x magnification; B, 400x magnification) The hypercellular region shows solidly arranged, forming microtubules of immature, poorly differentiated Sertoli cells and Sertoli cells with mucinous degeneration similar to signet ring cells. Poorly differentiated Leydig cells are cells between Sertoli cells. (C, 20x magnification; D, 40x magnification) The hypocellular region shows dominant mucinous degeneration Sertoli cells are shaped like signet ring cells. Atypical Leydig cells.

Based on Meyer's classification of differentiation, the tumors in both ovaries were diagnosed as high-grade Sertoli–Leydig cell tumors.14 The presence of heterologous elements, including mucinous differentiation and signet-ring-like cells, raised differential diagnoses of metastatic gastrointestinal adenocarcinoma (Krukenberg tumor), high-grade serous carcinoma, and signet-ring stromal tumors. Immunohistochemical analysis showed strong positivity for calretinin (cytoplasmic and nuclear) and inhibin (cytoplasmic), while epithelial membrane antigen (EMA) and cytokeratin (CK) were negative in all tumor cells, supporting the diagnosis of Sertoli–Leydig cell tumor.

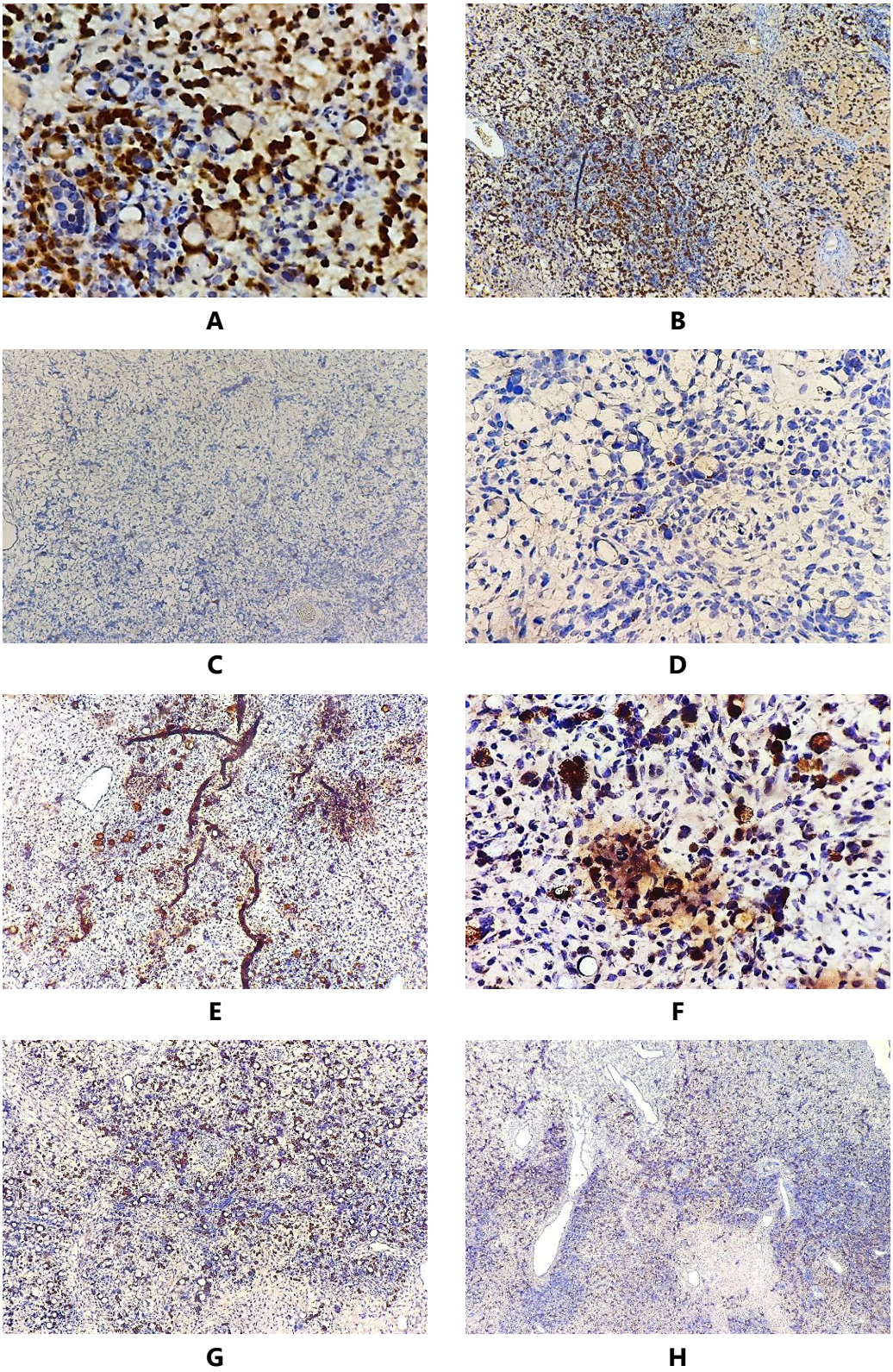


Figure 4. (A; B) IHC staining of calretinin has strong positivity in cytoplasmic and nuclei of Leydig cells in solid areas with a faintly staining fraction of Sertoli cells in the microtubule. (C; D) CK 7 negative staining. (E; F) EMA negative staining. (G; H) Inhibin cytoplasmic staining of Sertoli cell with an additional small contribution from Leydig cell.

One month later, the patient, classified as stage IIIC ovarian tumor, underwent common bile duct exploration and cholecystectomy. Histopathological examination of the gallbladder revealed tumor cell nests within the muscularis propria. The tumor cells were pleomorphic, with clear cytoplasm, irregular oval nuclei, and coarse chromatin. These findings were consistent with metastatic involvement, most likely originating from the ovarian tumor.

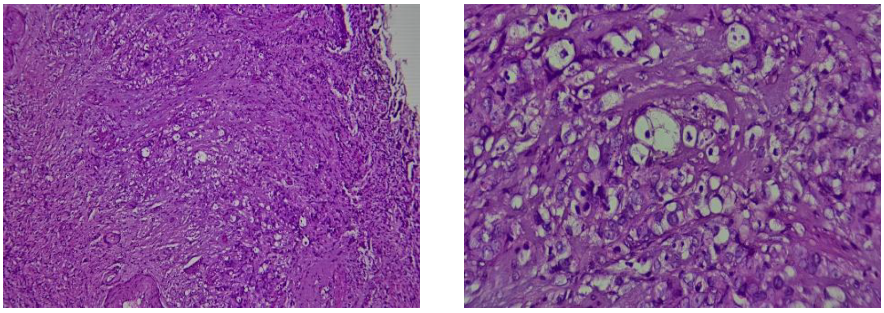


Figure 5. Secondary tumors of the gallbladder. The histopathological picture is the same as the SLCT of the ovary.

DISCUSSION

SLCT patients often exhibit endocrine manifestations, predominantly androgenic signs such as menstrual irregularities (oligomenorrhea or amenorrhea), hirsutism, voice deepening, clitoromegaly, prominent laryngeal features, and elevated serum testosterone prior to surgery.[17] Estrogenic manifestations, namely post-menopausal bleeding.[8,10,12] Some show non-hormonal symptoms in more than 50% of patients, namely abdominal pain, abdominal distension, and acute and severe abdominal pain due to tumor rupture.[11,18] Carcinoembryonic antigen (CEA) and CA-125 levels are at normal levels,(19) in SCLT high grade CA-125 levels increases to 190 U/L.[20]

Bilateral ovarian SLCT occurs in only 1.5% - 2% of all SLCT cases.[21] A literature study of bilateral ovarian SLCT cases obtained five cases of patients with [22–26] one patient over 50 years old and four patients aged 17-30 years. All cases show non-hormonal symptoms with the most common symptoms being abdominal mass accompanied by pain and abdominal distension. Laboratory tests showed that Alpha-fetoprotein (AFP) levels were increased (27.05 µg/L), while lactate dehydrogenase (LDH) levels (190.70 U/L) and CA 125 (4.78 U/mL) were normal or slightly increased (50.9 IU/mL). Ultrasound and MRI examinations show solid tumors, solid tumors with small cysts. Two cases of bilateral ovarian tumors the diameter of tumor up to 20 cm, multicystic, extending until entering the abdomen to reach the liver, gallbladder in the right region, and small intestine curve to the minor curvature of the stomach in the left region, to the right kidney in the posterior region of the abdomen, without evidence of metastases so the stage is classified as IC. One case of metastases was reported on omentum. Histopathology shows a population of two types of cells, the first is a small cell Sertoli cell, a cytoplasm, an oval round nucleus forming a solid nest, creeping and tubule, separated by fibrous connective tissue stroma, among which the second cell is a pale clear cytoplasmic cell with clustered vacuoles. Some cells exhibit the morphology of a signet ring cell—histopathology of intermediate to poor degree of differentiation.

The prognosis of SLCT primarily is primarily determined by the tumor stage and the degree of cellular differentiation.[21] SLCT is classified into low grade, intermediate grade, high grade, retiform variant, and those exhibiting heterologous elements. Assessment of differentiation relies on the extent of tubular formation by Sertoli cells, which progressively diminishes as the tumor becomes less differentiated. In contrast, poorly differentiated tumors demonstrate an increased proportion of primitive gonadal stroma, while the presence of Leydig cells correspondingly declines with worsening differentiation. Moderately and poorly differentiated SLCT may exhibit primitive gonadal stroma along with heterologous elements such as mucin-secreting epithelium, striated muscles fibers, cartilage, hepatoid tissue, and less frequently, neuroectodermal elements. These features represent characteristic morphological patterns associated with advance differentiation grades.[20,27] SLCT generally has a

moderate and poor degree of differentiation, 20% of which have glands as heterologous elements most of the glands or cysts coated with the gaster or intestinal epithelium are well or moderately differentiated.[19] Immunohistochemistry is a critical tool for diagnosing poorly differentiated SLCT. Typically, Sertoli and Leydig cells exhibit positive staining for inhibin and beta-catenin, which aids confirming the diagnosis. The expression patterns of inhibin and beta-catenin in SLCT help a pathologist to accurately diagnose and classify tumors in the Wnt signaling pathway and provide specific differentiation from other ovarian neoplasms.[27]

SLCT with moderate or poor differentiation is difficult to distinguish from stem cell tumors, serous neoplasms, carcinomas, and the rare primary Wilms tumors of the ovary. Histologically, the tubular structures in moderately differentiated SLCT differ from those seen in stem cell tumors. In poorly differentiated SLCT, the stromal component often shows a sarcomatoid pattern, whereas stem cell tumors typically display a thecoma-like background. Characterized by occurrence of heterologous components or retiform architectural patterns leads to SLCT. When both epithelial and stromal elements are present in SLCT, raising suspicion for carcinoma, the typical clinical features – usually occurring in younger patients and characterized by androgenic manifestations –can provide important diagnosis clue. Carcinosarcoma shows a negative inhibin staining and a positive EMA. The heterologous hepatoid element shows an eosinophilic granular cytoplasmic cell and a nucleus in the middle, resembling a Leydig cell. Leydig-cell can be distinguished to heterologous elements by immunohistochemistry panels examination of Melan-A, keratin, AFP, vimentin, inhibin, HEPAR-1, and arginase. Leydig cells exhibit inhibin positivity with absence of HEPAR-1 and arginase expression, whereas mucinous epithelium components demonstrate CK7 and CK20 reactivity. Definitive diagnosis often require a correlation of clinical context, histomorphology, and immunohistochemistry results.[27]

Sertoli–Leydig cell tumor (SLCT) commonly presents with endocrine manifestations, predominantly androgenic features such as menstrual irregularities (oligomenorrhea or amenorrhea), hirsutism, voice deepening, clitoromegaly, prominent laryngeal features, and elevated serum testosterone levels.[17] Estrogenic manifestations, including postmenopausal bleeding, have also been reported.8,10,12 However, more than 50% of patients may present with non-hormonal symptoms, such as abdominal pain, abdominal distension, or acute abdomen due to tumor rupture.[11,18] Tumor markers such as carcinoembryonic antigen (CEA) and CA-125 are typically within normal limits,[19] although elevated CA-125 levels have been reported in high-grade SLCT, reaching up to 190 U/L.[20]

Bilateral ovarian SLCT is rare, occurring in only 1.5%–2% of cases.[21] A literature review identified five reported cases,[22-26] including one patient over 50 years old and four patients aged 17–30 years. Most cases presented with non-hormonal symptoms, particularly abdominal mass, pain, and distension. Laboratory findings showed elevated Alpha-Fetoprotein (AFP) levels, while Lactate Dehydrogenase (LDH) and CA-125 were normal or only slightly increased. Imaging studies (ultrasound and MRI) typically demonstrated solid or mixed solid-cystic tumors. Some cases reported large bilateral tumors (up to 20 cm), extending within the abdominal cavity without evidence of metastasis, corresponding to stage IC disease, although omental metastasis has been described in one case. Histopathological findings commonly revealed two cell populations: Sertoli cells forming solid nests and tubules within fibrous stroma, and Leydig cells with clear cytoplasm; some cases also demonstrated signet-ring cell morphology and intermediate to poor differentiation.

The prognosis of SLCT is primarily determined by tumor stage and degree of differentiation.[21] SLCTs are classified into well moderately and poorly differentiated types, as well as retiform variants and those with heterologous elements. As differentiation decreases, tubular formation by Sertoli cells diminishes, while primitive gonadal stroma becomes more prominent and Leydig cells less conspicuous. Moderately and poorly differentiated tumors may exhibit heterologous elements, including mucin-secreting epithelium, skeletal muscle, cartilage, hepatoid tissue, and, rarely, neuroectodermal components.[20,27] Approximately 20% of SLCTs contain glandular heterologous elements, often lined by gastrointestinal-type epithelium.[19]

Immunohistochemistry plays a crucial role in diagnosing poorly differentiated SLCT. Tumor cells typically express inhibin and β-catenin, which assist in confirming the diagnosis and differentiating SLCT from other ovarian neoplasms.[27] Differential diagnoses include

germ cell tumors, high-grade serous carcinoma, metastatic adenocarcinoma, and rare primary ovarian Wilms tumor. Histologically, poorly differentiated SLCT may show sarcomatoid stroma, while germ cell tumors often exhibit a thecoma-like background. Carcinosarcomas are typically inhibin-negative and EMA-positive. Leydig cells can be distinguished from heterologous elements using immunohistochemical markers such as Melan-A, keratin, AFP, vimentin, inhibin, HEPAR-1, and arginase. Leydig cells show inhibin positivity with negative HEPAR-1 and arginase expression, whereas mucinous epithelial components are positive for CK7 and CK20. Ultimately, accurate diagnosis requires correlation of clinical features, histopathological findings, and immunohistochemical profiles.[27]

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CONCLUSION

This case of high-grade Sertoli–Leydig cell tumor (SLCT) represents a diagnostic challenge in histopathological evaluation due to the presence of heterologous elements within the tumor. This report adds to the limited literature on high-grade SLCT, particularly in cases where the tumor remains confined to the ovaries despite its large size. In this patient, jaundice was the initial clinical presentation and was attributed to cholecystitis secondary to cholelithiasis. Histopathological examination of the gallbladder revealed tumor cell nests with a biphasic pattern similar to those observed in the ovarian tumors, suggesting metastatic involvement. To our knowledge, this is the first reported case of bilateral high-grade SLCT with metastasis to the gallbladder wall.

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